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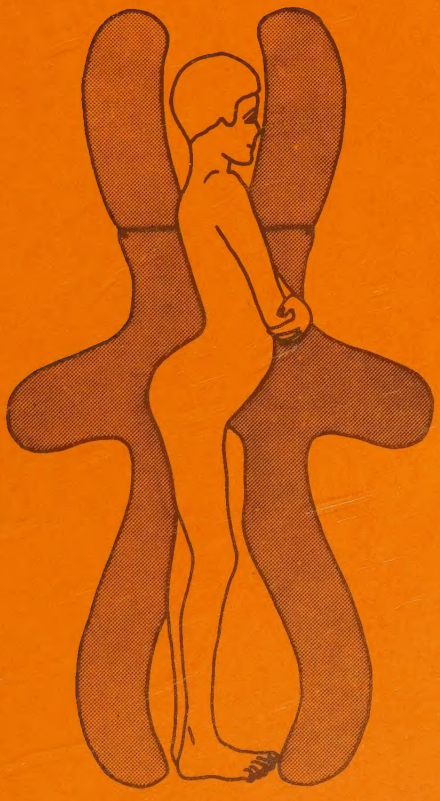
CONGENITAL DISLOCATION OF THE HIP

by

Nis Fredensborg



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TO SOPHUS VON ROSEN

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INTRODUCTION

Hippocrates (460-370 B.C.) probably gave the first description of congenital dislocation of the hip (CDH). He realized that the condition was sometimes present at birth and that prompt treatment was imperative. More than two millenia elapsed until this opinion was again advanced. Roser (1879), at a congress in Berlin, entreated his fellow-surgeons to test his 15-year-old theory that almost all cases of hip dislocation could be cured if diagnosed at birth and treated in abduction. He thought that the condition was induced by an abnormal fetal position, particularly in girls. Lorenz (1920), however, postulated that CDH was an acquired deformity caused by an inhibition of the growth of the acetabulum. He considered that treatment should be started at 2-3 years of age. The principles of Lorenz were dominant for many years. However, already Le Damany in 1912 had described the classical diagnostic sign of hip dislocation in the newborn child, "signe de ressaut", and in Italy early diagnosis and treatment were successfully introduced by Putti (1927) and Ortolani (1937), and gradually these principles were universally accepted

In Sweden these concepts were introduced in the late forties, the first data being presented by Palmén (1953) and von Rosen (1956). Throughout Sweden the principle of early diagnosis and treatment has been generally accepted for two decades although it has been applied with a varying degree of success in different parts of the country. This general acceptance has, in fact, been world-wide during the sixties and not until recently opposed by a number of investigators.

Former investigators have claimed that it is possible to diagnose all or almost all cases of CDH already in the newborn child and that with proper treatment these children will develop normal hips. These two statements have been challenged.

Wynne-Davies (1970) suggested that there are two types of CDH. In one type there is a congenital joint laxity causing unstable hips at birth. The other type, however, is due to a progressive acetabular dysplasia which leads to dislocation of the hip during early childhood. Walker (1971) supported this hypothesis. He found that many children presented with dislocated hips later in life even if the hips had been stable at birth. Williamson (1972) was of the opinion that the diagnostic procedure carried out at the time of birth can be a failure and also stated that early treatment may not always prevent dislocation. In fact, early treatment has by some investigators been characterised as not only unsuccessful but also dangerous (Mears 1974) and surgical reduction is suggested as the best and least harmful method in that reduction and splinting cause avascular necrosis of the femoral head in the newborn child.

In Norway the application of early diagnosis and treatment was discouraging (Bjerkreim 1974). In spite of a considerable overtreatment including possibly five times the number of expected cases, most cases are being missed. Not to mention that many of the treated cases, whether originally dislocated or not,have developed abnormal hips.

These reports are in contrast to our experience in Malmö as reported by von Rosen (1970). It is possible to diagnose almost all children with CDH already at birth and treatment in the von Rosen splint will result in hips that are normal in all respects. In view of this new development we have decided to re-examine our cases and to survey our position with regard to the diagnostic procedure.

One large group of children, born in the city of Malmö during the years 1956-1972 were therefore selected for re-examination. These children were examined with regard to the effect of the diagnostic procedure. A subset of these children, namely those diagnosed in the years 1956-1964, were also re-examined with a clinical and radiological follow-up in order to study the hips at a later stage of development when the final shape of the joint may be observed or at least predicted with some accuracy.

In addition, certain data have been collected which may add to our knowledge of the pathogenesis of CDH, namely, studies of joint laxity and of connective tissue composition.

Standard statistical methods were applied (Snedecor and Cochran 1967).

PART I

THE EFFECT OF EARLY DIAGNOSIS OF CONGENITAL DISLOCATION OF THE HIP

Some reports have been very discouraging with regard to the possibility of early diagnosis of CDH. In some instances (Williamson 1972 and Bjerkreim 1974) the number of missed cases is so large that most of the expected cases are probably included in this group, whereas a group, many times larger, which presumably would not have developed hip dislocation, is diagnosed and treated. This is indeed a serious matter, if one also takes into account the discouraging reports on treatment with damage to the femoral head caused by immobilazation.

These data could be interpreted to mean that by early diagnosis most cases of CDH are being missed whereas a number of normal children are splinted with resulting damage to their hips.

The aim of the present investigation was to study the variations in early diagnosis of CDH with regard to its efficiency and to the number of cases being treated on the basis of this diagnostic procedure.

MATERIAL

During the years 1956-1972 altogether 58,759 children were born in the city of Malmö. All these children were or were supposed to be examined within the first few days of life with regard to CDH. During the first few years of this period, examination of newborn children was undertaken twice a week so that the time interval from birth to examination never exceeded four days. In most of the children, however, examination was undertaken on the day of or on the day after birth. Until 1963, the hips of the newborns were examined only once but during 1963-1972 the children were examined not only within 24 or possibly 48 hours but also on the day of discharge from the maternity ward.

Until the middle of 1963 the Ortolani sign was used as the criterion of CDH. After this date the method of Palmén (1957) was also introduced. This included the subluxation provocation test.

All the children were radiographed according to Andrén (1961[a] and 1962). The radiological criteria of CDH were one or more of the following:

1. The hips were examined in flexion and abduction and pushed together and pulled apart to demonstrate widening of the joint space and/or widening of the symphysis as a sign of dislocation and ligamental laxity (Figures VII:1 and VII:2).
2. In slight abduction and outward rotation the hips were forced outwards and upwards to a position of dislocation (Figure VII:3).
3. With straight legs and inward rotation the femora were forced upwards and especially in unilateral cases a distinct difference in upwards displacement was noted (Figure VII:4).
4. In 45 degrees of abduction and inward rotation the unstable hip was forced out of the socket (Figure VII:5).

The children were classified as CDH if one radiogram was obtained which obviously showed at least one hip in a dislocated position. In a number of cases, diagnosed clinically by the paediatrician, the radiograms were classified as negative for CDH. These children were nevertheless treated.

Over the years altogether 548 children, 430 girls and 118 boys, were diagnosed and treated (Tables I:1 and I:2).

Table I:1. The incidence of diagnosed and missed cases of CDH in Malmö during 1956-1972.

	Number	Incidence/1000
Total live birth	58,759	
Children with CDH	548	9.30
Missed cases	4	0.07

Table I:2. Distribution according to sex and side. Bilateral cases are grouped according to predominant instability.

	Bilateral			Unilateral		
	Left > right	Right > left	Equal	Left	Right	Total
Girls	83	68	108	100	71	430
Boys	19	14	22	43	20	118
Total	102	82	130	143	91	548

These children were classified according to the diagnostic procedure in the following four groups:

von Rosen material (1956-1964): The majority of these children were diagnosed by Doctor P. Selander in the Department of Paediatrics of the Malmö General Hospital and later treated by Doctor Sophus von Rosen in the Orthopaedic Department of the same hospital. In most instances the diagnosis was confirmed by a radiogram, obtained and evaluated by Doctor L. Andrén in the Department of Radiology. This particular subset of the material has been re-examined and will be presented in Part II of the present study.

Andrén positive (1965-1972): In these children the clinical diagnosis was confirmed by Doctor L. Andrèn who obtained and evaluated the radiograms in all instances.

Roentgen positive (1965-1972): Children with a positive radiogram, not evaluated by Andrèn.

Roentgen negative (1965-1972): Children classified as CDH by the paediatrician only. The radiograms were classified as negative but the children were nevertheless treated.

During the first years mainly three individuals were involved in the diagnostic procedure: Selander, Andrèn and von Rosen. In 1963 Selander retired and in 1965 von Rosen followed suit. After the retirement of the two department heads the responsibility for diagnosis of CDH was divided between several individuals both in the Department of Paediatrics and in the Department of Orthopaedics. In some time periods the responsibility was, in fact, not very well defined.

Of these children, 9 were classified as atypical

2 with arthrogryposis
2 with myelomeningocele
2 with cerebral palsy
1 with Bonnevie-Ullrichs syndrome
1 with trisomia
1 with multiple deformities. The child died within 3 months.

RESULTS

The incidence of diagnosis of CDH increased during the period of investigation. In 1964-1966 the increase was immense and reached a maximum of almost 2 per cent. Later on, the incidence decreased but still maintained a comparatively high level (Figure I:1)

The enormous increase in the mid-sixties appears to be related in time to the retirement of the two department heads. Also, in 1963, the subluxation provocation test was introduced.

Figure I:1. The incidence of CDH in Malmö 1956-1972.

In 4 children the diagnosis of CDH was missed at the initial examination:

1. 1960, a girl. Vertex presentation of a normal child. The hips were examined within 48 hours and considered to be normal. Unilateral CDH was diagnosed at 18 months when the mother had discovered that the child was limping.

2. 1963, a girl. Vertex presentation of a normal child. At examination within 48 hours the hips were classified as normal. At the age of 18 months unilateral CDH was diagnosed. The child had started to walk 1 month earlier and had been noticed limping

3. 1966, a girl, Vertex presentation of a normal child. The hips were examined within 48 hours and classified as normal. Five weeks later bilateral CDH was diagnosed at a Childrens Welfare Clinic.

4. 1971, a girl. Vertex presentation of a normal child. The hips were examined within 48 hours and at the time of discharge from the maternity ward. In both instances the hips were classified as normal. At the age of 14 months unilateral CDH was diagnosed when the child started to walk.

In none of these four children can the failure of diagnosis be explained retrospectively. They were all considered to be normal, healthy children and were handled according to the standard procedures.

DISCUSSION

The overall incidence of CDH during the years 1956-1972 was 9.3 per thousand. During the same time period the incidence of missed cases was 0.07 per thousand. Severin (1956) calculated the total incidence of CDH in Sweden to 0.9 per thousand. von Rosen (1961) found, for Malmö, the incidence to be 1.7 per thousand. This indicates that at present five times as many children are being treated for CDH as would otherwise have developed a manifest dislocation. The incidence of diagnosis is well in agreement with data from other parts of Sweden (Palmén and von Rosen 1975). Also incidence peaks such as were observed in the city of Malmö in 1966 have been found elsewhere. In a recent report Artz et al. (1975) found an incidence of 14 per thousand (New York City). A similar order of magnitude was presented by Barlow (1966) (Manchester) and MacKenzie (1972) (Aberdeen)

In the present study the incidence of missed cases is extremely low. However, similar success for diagnostic routines have been presented by Barlow (1966) (Manchester), Finlay et al. (1967) (Middlesex), Mitchell (1972) (Edinburgh) and Artz et al. (1975) (New York City). Obviously it is a common experience that CDH can almost always be diagnosed in the newborn child. Recent reports, however, present quite different data. Williamson (1972) demonstrated how one unit of the maternity ward in Belfast missed 2 per thousand of the children with CDH in one part of the population whereas in another sample of the same population the total incidence was less than that. This indicates that all cases

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were missed. Similar confusing data have been presented by Bjerkreim (1974). More than 7 per thousand of the newborns in southern Norway are diagnosed in the neonatal period and treated for CDH. Nevertheless, 0.8 per thousand or 2/3 of the expected incidence in Norway before early diagnosis present later with manifest dislocation.

It is at the present time not possible to evaluate the ethnical differences in this disease. Wynne-Davies (1970), for instance, claims that CDH may develop later in life in originally normal hips. This is certainly not the experience in Malmö nor obviously in many other centres. Probably experience in the diagnostic procedure is of importance as well as the time chosen for examination, which should be within the first two days of life. The importance of an experienced investigator has been demonstrated by Moore (1974).

A simple but firm organisation for the diagnostic procedure as instituted by Selander is probably of great value. The lack of organisation or rather the distribution of responsibility which took place during the course of the period under examination in the present study did, however, not introduce a greater number of missed cases but rather the treatment of a large number of children, some of whom of necessity must have been normal.

The radiological examination of the newborn implies a confirmation of the clinical diagnosis. In some cases, in which a negative radiological diagnosis had been submitted, the children were nevertheless treated. The advantage of a radiological diagnosis has not been generally accepted and the danger of relying on a negative radiogram was demonstrated by Palmén (1970). If, however, the radiograms are obtained by the techniques described by Andrén (1962) the risk of false negatives is probably negligible. In the present study those children who had a negative radiogram and who were not treated, were not re-investigated. To our knowledge none of these children later on was found to have CDH.

PART II

THE RESULTS OF EARLY TREATMENT OF CONGENITAL DISLOCATION OF THE HIP

It has been established that in the city of Malmö almost all cases of CDH can be dagnosed at the time of birth. The aim of the present study was to investigate whether or not a normal development of the hip is to be expected provided that treatment is commenced within the first few days.

MATERIAL

During the years 1956-1964 a total of 30,280 live births were recorded in the city of Malmö, 99.5 per cent of which took place at the maternity ward of the Malmö General Hospital. All the children were examined by a trained paediatrician. During the first part of the time period under study, examination of the hip joints of the newborns was undertaken once or twice a week, but later on daily. The children were examined by the method described by Ortolani (1937). The diagnosis was confirmed by radiological examination according to the methods of Andrén (Andrén and von Rosen 1958). From 1959 onwards the signs of pelvic instability were also considered (Andrén 1960 and 1961[a]). In most instances the radiological examination was carried out by Doctor Andrén personally. The diagnosis of CDH was therefore based not only on the clinical but also on the radiological finding. It must be concluded that in the cases comprised in the present study the hip was dislocatable or pathologically unstable at least on one occasion, namely, at the time of the radiological examination. In a few cases with an incomplete radiological examination, the diagnosis of CDH was based on the clinical examination only which in these cases was confirmed by two experienced physicians and therefore probably beyond doubt.

Of 119 cases diagnosed within the first few days of life and treated in the von Rosen splint, there was one child with arthrogryposis, one with myelomeningocele, one with Bonnevie-Ullrichs syndrome and one with cerebral palsy. Another child died later on of cardiac failure and three had emigrated leaving 111 typical cases of CDH for the follow-up examination. These cases were distributed as demonstrated in Table II:1.

Table II:1. Distribution according to sex and side.
Bilateral cases are grouped according to predominant instability.

	Bilateral			Unilateral		Total
	Left > Right	Right > Left	Equal	Left	Right	
Girls	26	13	19	21	14	93
Boys	4	4	1	8	1	18
Total	30	17	20	29	15	111

The period of treatment in the von Rosen splint varied between 2 and 20 weeks with an average of 10 weeks. In the very beginning of the time period under study the duration of splinting was 3 months. Later on, in late 1959 and early 1960, some experimenting was undertaken with successive shortening of the period of fixation. One girl was splinted for 2 weeks only and re-dislocated. Subsequently the 3 months policy was resumed except for a short period in 1963 when, again, some experimenting with shorter fixation was undertaken, this time without failures.

With some individual variation the children were seen in the Department of Orthopaedic Surgery within one week after birth and put in the von Rosen splint. Thereafter they were seen by the surgeon every 2 - 3 weeks until the splint was removed. Once a week the children were bathed in the outpatient department by an experienced nurse. The mother was not allowed to remove the splint at all. After removing the splint, with some individual variation, the children were seen at 6 and 12 months and later on at 3, 5 and 7 years of age. Radiological re-examinations were performed at the same intervals as the clinical follow-up examinations.

The initial treatment included two failures. One is the above-mentioned child, who re-dislocated after 2 weeks of splinting. The failure was discovered after another 2 weeks. Plaster immobilization in Lorenz' first position was applied and had to be maintained for 10 months owing to persistent instability. Later this child also developed a pubertas praecox but hip development was normal (Figure VII:6).

In another girl the initial treatment was uneventful. The radiological appearance of both hips at 2 months was normal. At 7 months the acetabulum could be suspected to be somewhat steep on one side and at 12 months subluxation was visible (Figure VII:7). This child was treated in a special abduction splint and later on developed normal hips (Figure VII:8).

METHODS

Clinical examination: The clinical examination was undertaken at an average of 10.3 years (8 - 16 years). The following variables were recorded: walking age, hip symptoms, gluteal insufficiency and range of motion.

Radiological examination: [1] Radiological examination was in most instances undertaken at the time of the follow-up. In a few cases films taken one year prior to the follow-up examination were used. In no instances was the child younger than 8 at the time of the radiological examination. The examination included an antero-posterior film of the pelvis including both hips and the upper end of the femora. The hips were in a position of maximum inward rotation. In all but 27, the examination included the so-called Lauenstein position with the hips in flexion, abduction and outward rotation. The radiograms were taken with a film-focal distance of 125 centimeters. The general configuration of both hips was noted. The centre-edge angle (CE angle) according to Wiberg (1939) was measured (Figure II:1).

In order to examine the roundness of the femoral head a new index was designed, expressing in per cent the relationship of the maximum width of the

1) The radiological examination was performed by Doctor L. Andrén, Roent-

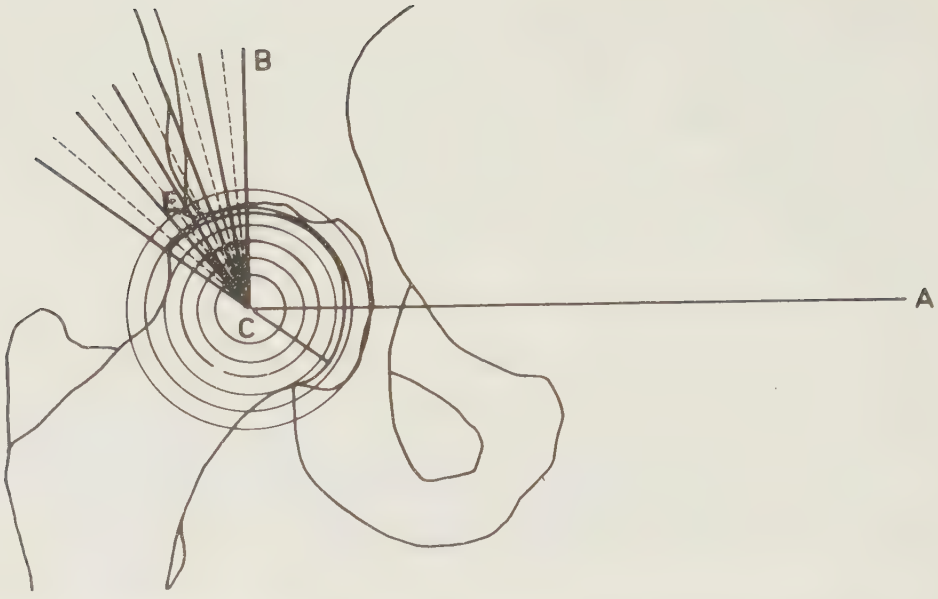

Figure II:1. The CE angle according to Wiberg (1939). From the centre (C) of the femoral head a line (C-A) is drawn through the centre of the head of the opposite side. Perpendicularly to this line and through the centre (C) the line (C-B) is raised. The CE angle denotes the angle between the line C-B and a line from C to the acetabular edge (E).

head and the maximum height of the head which could be measured perpendicularly to the line denoting the width (Figure II:2). This variable is referred to as the spherical index.

The initial films were reviewed not only with regard to the signs of CDH in the newborn but also with regard to the size of the bone nuclei of the capital epiphyses, when these became visible.

For the purpose of comparison, particularly with respect to the CE angle and the spherical index, films of 222 children with the same age and sex distribution were studied in the same manner. These children had all been radiographed after 1950 for reasons such as trauma or transient hip pain. The method of obtaining pelvic radiograms had not been systematically changed during this time period except for the fact that the rotational position of the control children could not be established in retrospect.

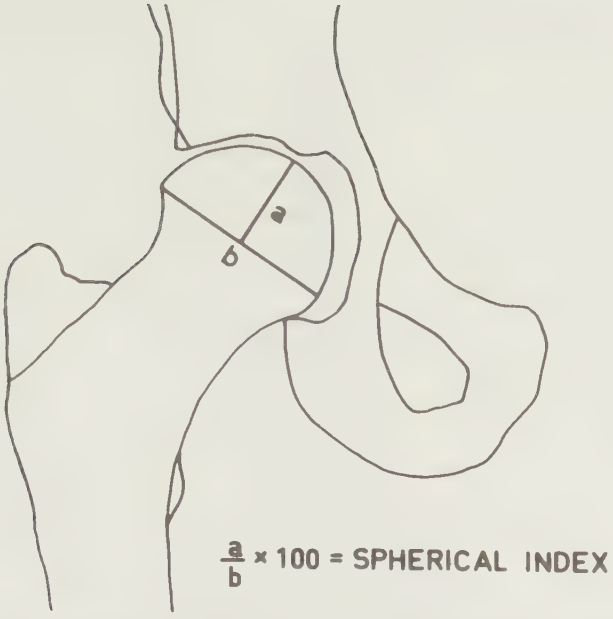

Figure II:2. The spherical index. The line (b) denotes the maximum width of the femoral head and (a) the maximum height measured from this line.

RESULTS

Clinical examination

Walking age: Children with CDH in this series started to walk at the age of 13.1 ± 2.2 months. This might be compared with the average normal walking age stated by Hindley et al. (1966). Compared with the group of children from Stockholm, who started to walk at 12.5 ± 1.2 months, there is a delay of 2 weeks. This difference is significant ($0.01 > P > 0.001$). There is one distinct difference in obtaining the data for the two groups in that the data for the control children were part of a longitudinal study whereas the data for the present study had to be obtained in retrospect.

Hip symptoms: At the time of the follow-up none of the children complained of hip symptoms and in no instances could there be established any disability in conjunction with daily life or sports activities.

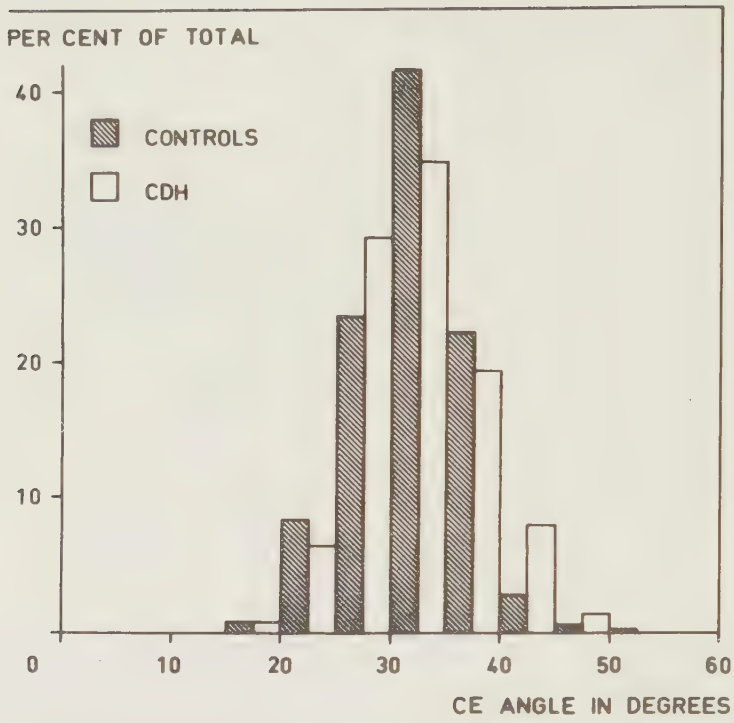

Figure II:3. The CE angle in 222 CDH hips compared with 444 controls.

Gluteal insufficiency: No case of gluteal insufficiency was recorded.

Range of motion: The range of hip motion was measured with a goniometer and was in all cases within the range of normal (American Academy of Orthopaedic Surgeons 1965).

Radiological examination

Visual impression: The general configuration was normal in all but two cases (cf. case reports).

CE angle: The CE angles compared with the controls were of an identical distribution (Figure II:3). This indicates a normal development of the acetabulum. If, however, the CE angle was studied in relation to age there was a significant difference. The CDH girls appeared to have a slightly lower angle at an early age and later on developed the same or even a greater angle, that is a less steep acetabulum (Figure II:4).

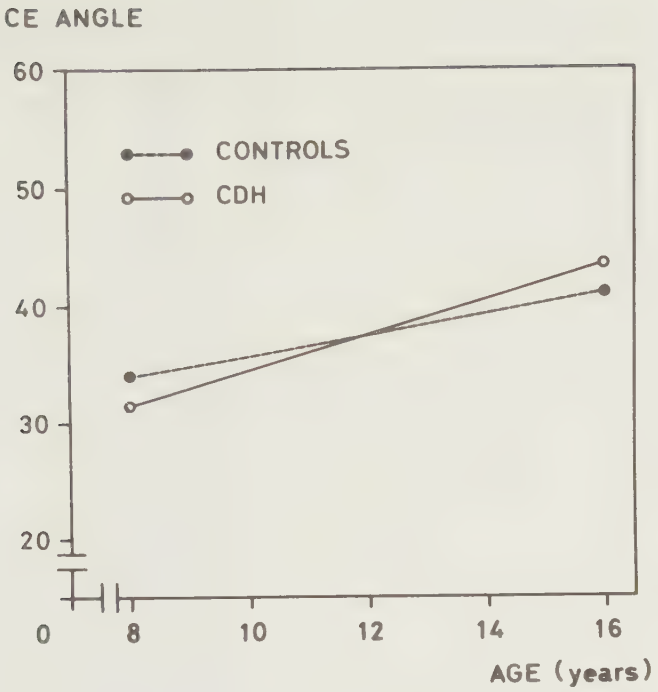

Figure II:4. The relationship between age and CE angle in CDH and control girls. There was a significant positive correlation ($P < 0.001$). The slope was significantly steeper in CDH girls ($0.01 > P > 0.001$).

Spherical index: The spherical index also was identical to that of the control population (Figure II:5). This indicates a normal development of the femoral head in children with CDH. Also the spherical index increased significantly with age ($P < 0.001$) indicating that the capital epiphyses is somewhat flatter in young children. The development was in this respect identical in CDH girls and controls.

There was no side to side difference of the CE angle or the spherical index between the stable and unstable hip in unilateral cases. These anthropological measurements indicate that children with CDH develop a normal hip with a deep acetabulum and a round femoral head.

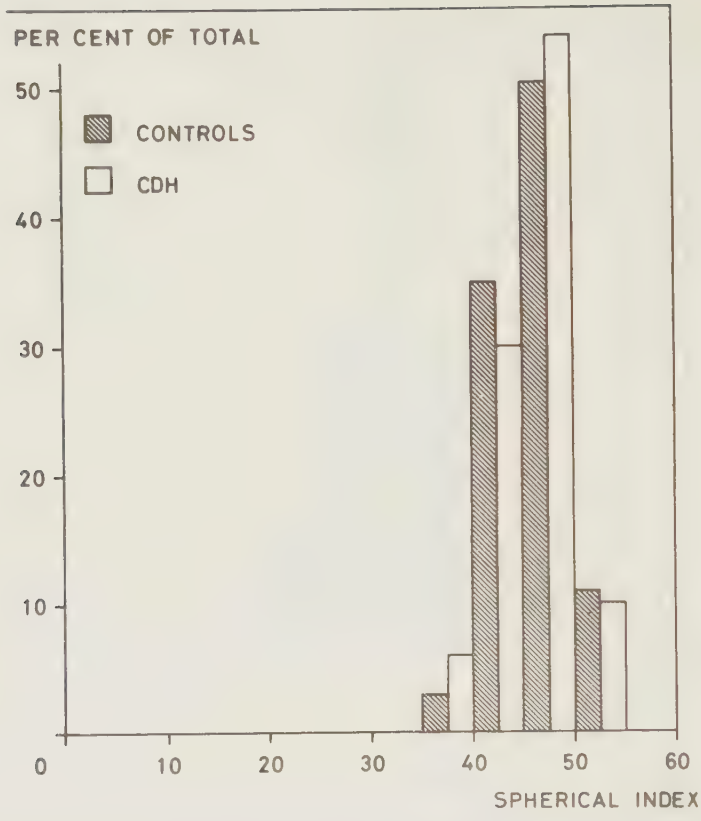

Figure II:5. The spherical index in 222 CDH hips compared with 444 controls.

Capital epiphyses: The capital epiphyses were found to be significantly smaller in the dislocated hip in unilateral cases (Table II:2) and in the less stable hip in bilateral cases with side to side difference (Table II:3). There was no right-left difference in bilateral cases without difference in stability between the two hips (Table II:4).

Deviating cases (case reports)

Case 1. Girl, who was initially treated without complication and showing an initially normal development of the hip joint. At the time of follow-up at age 10, however, the shelf was steeper in the originally dislocated joint than in the undislocated. Nevertheless, the CE angle is within normal limits (15 degrees according to Wiberg 1953) and the femoral head is not displaced (Figure VII:9).

Table II:2. Side difference (mm) between the capital epiphyses (dislocated minus stable) in unilateral CDH.

	Left minus right	Right minus left
Number	29	15
Average	-1.00	-0.87
S.D.	1.46	1.19
	$P < 0.001$	$0.02 > P > 0.01$

Table II:3. Side difference (mm) between the capital epiphyses (less stable minus more stable) in bilateral CDH with unequal instability.

Number	47
Average	-0.64
S.D.	1.52
	$0.01 > P > 0.001$

Table II:4. Side difference (mm) between the capital epiphyses (right minus left) in bilateral CDH with equal instability.

Number	20
Average	-0.05
S.D.	1.32
	$P > 0.5$

Case 2. Girl, who was originally treated without complication and who developed a normal hip joint. A radiogram at age 1, however, disclosed a small cyst and later on, (Figures VII:10 and VII:11), the roentgenological appearance of avascular necrosis was noted. This girl later on developed a completely normal hip except that the femoral head and neck were slightly larger (Figure VII:12). In this case both hips were originally dislocated. This girl had no physical findings and no symptoms at the time of follow-up.

DISCUSSION

Weissman and Salama (1966 and 1969) found that a considerable number of the children treated in the von Rosen splint or a Freika-like pillow after determination of treatment at 3 months were still dislocated. Williamson (1972) maintained that early treatment did not always prevent dislocation.

Bjerkreim (1974) found that after treatment with the Freika pillow, 30 per cent of the cases needed further treatment such as plaster fixation or surgery. Of those treated with the Freika pillow alone, acetabular dysplasia was observed in 12 per cent.

Not only failure but also a dangerously high complication rate in early treatment has been reported. Williamson and Carter (1960) found signs of avascular necrosis in one fourth of the children who were reduced by manipulation under anaesthesia. Similarly MacKenzie et al. (1960) found avascular necrosis in one fourth of the cases after manipulation. The fact that they were manipulated, however, suggests that the children were not newborn but that the diagnosis was late and that the reduction was done after abduction contracture of the hip had been established. Mitchell (1972) described two cases of transient deformity of the hip after splint fixation, possibly avascular necrosis. Felländer et al. (1970) described osseous changes in the acetabulum and in the femoral epiphyses in 40 per cent of their cases. These cases were treated as newborns in a modified von Rosen splint with rigid fixation. The first report of a case of avascular necrosis in its usual sense or at least a radiological finding which is consistent with early avascular necrosis is, in fact, in this paper, if only children splinted within the first few days of life are included. Mears (1974) suggests that avascular necrosis is such a common and harmful complication to treatment of CDH that the newborn child should not be subjected to this treatment but rather operated on. In a recent editorial (Lancet 1974) a warning is issued against splint treatment of CDH based on data of avascular necrosis following plaster treatment. We maintain that avascular necrosis is a rare, benign complication to early treatment providing that early treatment is defined

as treatment commenced in the newborn. This point of view is shared by other investigators (Ponseti 1966).

The duration of treatment varied considerably in the present study. It has been suggested that 6 weeks of treatment is adequate in all cases (Emnéus and Undeland 1970) and also that only dislocated (Barlow 1966) or dislocatable but not unstable hips require treatment (Finlay et al. 1967). Since treatment has caused no harm to the development of the hip joint of the children in the present study we continue to treat every child for 3 months irrespective of the degree of instability.

It is a common experience in follow-up studies of CDH that the functional result is better than the anatomical (Severin 1941). Consequently, an absolutely normal clinical follow-up was found as might be expected.

In the present study the hip joints of the children at the time of the follow-up were undistinguishable from the hips of normal children. One single child could possibly be suspected of having an abnormal hip development, but, on the other hand, the shape of the hip of this girl may not necessarily be related to the fact that she was born with an unstable hip.

PART III

ADDITIONAL OBSERVATIONS IN CHILDREN WITH CONGENITAL DISLOCATION OF THE HIP

The case histories and the vital statistics of the children included in the study on the effect of early treatment included some additional information which will be presented briefly.

MATERIAL AND METHODS

The same children who were re-investigated and who were treated as newborns for CDH with the von Rosen splint were included in the study of the additional data. The information was collected from the maternity records and from interviews with the child and parents.

RESULTS

Fetal presentation: Thirty-one per cent of the girls and 33 per cent of the boys were born in the breech presentation as compared to the average 5.7 per cent in Malmö. Of the children born in the breech presentation, 78 per cent were first born. One girl was delivered by Caesarean section. One girl was number one of a pair of dizygotic twins (number two, a boy, was unaffected).

Birth rank: Sixty per cent of the patients were first born whereas the average for Malmö is 45 per cent. The difference was significant ($0.01 > P > 0.001$).

Birth weight: The birth weight in CDH girls was $3,457 \pm 530$ g. In a random control sample from Malmö, the birth weight of 100 girls was $3,352 \pm 465$ g. The difference was not significant ($0.2 > P > 0.1$)

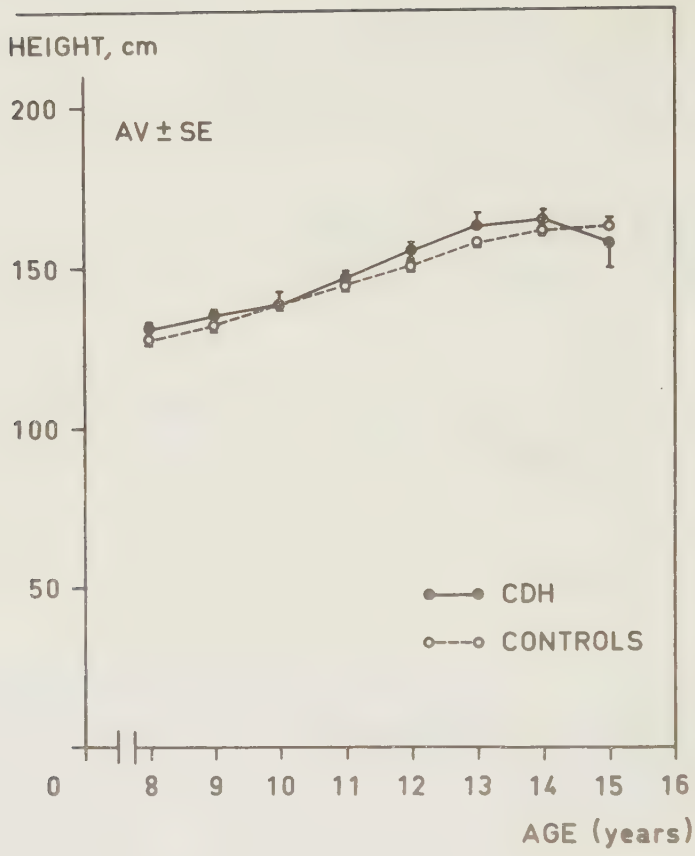

Figure III:1. The height of 91 CDH girls compared with 554 controls.

Height and weight: In this study only girls were included. The height and the weight were compared with a control sample (Karlberg et al. 1975). There were significant differences between CDH girls and controls in that CDH girls were somewhat taller and somewhat heavier (Figures III:1 and III:2).

Menarcheal age: The menarcheal age in the oldest 19 girls in the CDH series was 12.3 ± 1.4. This means, however, that in most girls the menarcheal age is not known and that a late menarche must be expected in some of the remaining individuals. Therefore, the difference between these data and data on menarcheal age from a contemporary Scandinavian material (Andersen 1968), 13.3 ± 1.3 years, is not necessarily significant.

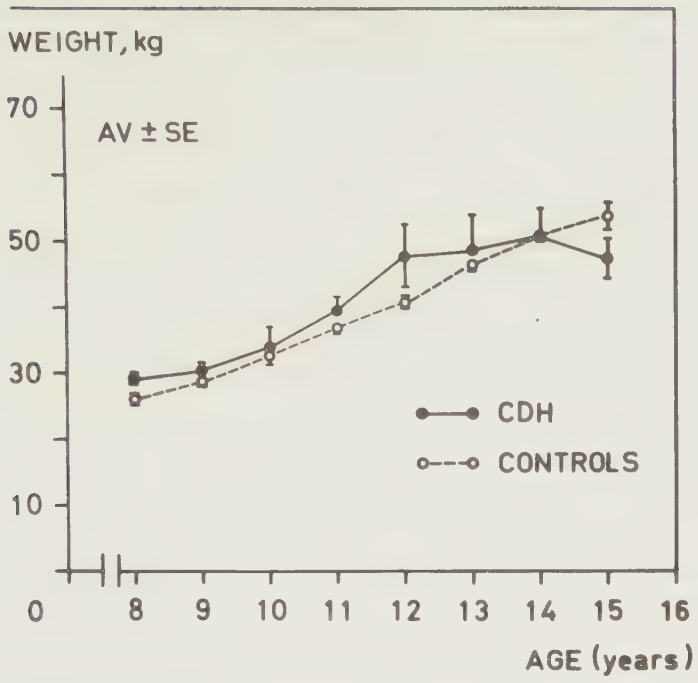

Figure III:2. The weight of 91 CDH girls compared with 554 controls.

Seasonal variation: The incidence of diagnosed CDH in newborns was higher in August to November but the deviation in this series was not significant.

Heredity: An attempt was made to estimate the prevalence of CDH in the families of the CDH children. Only an interview with one of the parents was made. A number of children of the same generation had been treated after early diagnosis in these families: Five siblings, four first cousins and five second cousins. In the previous generation CDH was known in one mother and suspected in a few more cases who had obviously had hip conditions. The data do not permit any further conclusions about the heredity of CDH.

Concomitant anomalies: During the years 1956-1964 a total of 121 children were diagnosed as having CDH. Among these a series of anomalies were recorded (Table III:1).

Table III:1. Concomitant anomalies in 121 children with CDH.

Anomaly	Number	
	Boys	Girls
Congenital heart failure	1	
Testes not descended	1	
Myelomeningocele		1
Arthrogryposis		1
Bonnevie-Ullrichs syndrome		1
Torticollis		1
Pubertas praecox		1
Inguinal hernia		
bilaterally		2
unilaterally	1	2

DISCUSSION

The sex ratio in this study, 5.2 - 1, is in agreement with that of earlier investigators. Idelberger (1951) reported a ratio of 5.4 - 1 based on 45,611 cases and Palmén (1961) a ratio of 4.1 - 1 in 894 neonatal cases and 5.8 - 1 in 1,486 mainly late-diagnosis cases. A similar difference in sex ratio between neonatal and late-diagnosis was reported by Wynne-Davies (1970) and Bjerkreim (1974). Breech presentation has been referred to as an environmental factor in the etiology of CDH (Record and Edwards 1958 and Andrén 1961[b]). Several investigators (Wilkinson 1963 and 1972, Carter and Wilkinson 1964[a] and Fettweis 1973) claimed that the risk of CDH is particularly increased in breech malposition. The association of breech presentation and birth rank was reported by Record and Edwards (1958), von Rosen (1959) and Andrén (1962). If, in the present series, cases born in the breech presentation are excluded, no primogeniture effect remains. This has also been reported by Bjerkreim and van der Hagen (1974).

A few investigators have observed deviations from the average in birth weight in CDH children (Record and Edwards 1958, Bjerkreim and van der Hagen 1974 and Artz et al. 1975). However, the investigators do not agree as to the direction of this deviation. In the present study the difference was not significant.

As for the concomitant conditions the findings in this study are in agreement with previous investigators. Concomitant malformations has been reported as occurring in CDH children to an increased degree by Record and Edwards (1958), Wynne-Davies (1970) and Bjerkreim and van der Hagen (1974). The data of the present study are not conclusive but taken together with the observations of other investigators it seems to indicate such an increased prevalence. The prevalence of inguinal hernia is of special interest. Inguinal hernia is extremely rare in girls (Knox 1959). In this series the expected number is very close to 0 but 4 cases of hernia was observed. Similar small increases in the prevalence of hernia in CDH children have been observed by Wynne-Davies (1970) and Bjerkreim and van der Hagen (1974). Taken together these studies indicate a significantly increased risk of inguinal hernia in CDH girls.

Most of the data on CDH children in the present study are in agreement with those of other investigators and merely confirm these findings as well as the identity of the sample. One observation, however, has not been previously presented, namely, the fact that CDH girls are somewhat taller and somewhat heavier than controls. These findings cannot be interpreted at present but might be caused by an earlier maturation in girls born with CDH.

PART IV

OVERDIAGNOSIS OF CONGENITAL DISLOCATION OF THE HIP

It has previously been demonstrated by many investigators (Putti 1933, Ortolani 1951, Coleman 1956, Palmén 1961, Barlow 1962, Emnéus 1966, Finlay et al. 1967, von Rosen 1968 and Bjerkreim 1974) that the number of hips diagnosed and treated in a given population when the method of early diagnosis is introduced is several times larger than the number of manifest dislocation diagnosed at older age. A reasonable question is whether these additional cases are all normal, healthy children or whether the diagnostic procedure in fact reveals some abnormality possibly related to CDH. It was decided to study the prevalence of some variables typical for or known in CDH, in a group of patients in whom the diagnosis was established with various degrees of certainty. At the same time this might provide a crude estimate of the validity of the diagnostic procedure.

MATERIAL AND METHODS

During the years 1956-1972 altogether 548 cases of congenital dislocation of the hip were diagnosed in the city of Malmö. During these 17 years the expected number based on the approximate incidence of 1 per thousand in the population should have been about 60 cases. This leaves almost 500 cases of whom it is not known to what extent they belong to the CDH group.

The cases were subdivided into 4 groups according to the method of diagnosis (cf. Part I).

It is assumed that the von Rosen material has the highest incidence of true CDH based on the fact that two experienced physicians and one experienced radiologist agreed on the diagnosis. The Andrén material is second in that the diagnosis was confirmed in all instances by the same radiologist. The roentgen diagnosis, whoever is responsible for it, may add some information towards the diagnosis and is given the third rank. Cases which are diagnosed by the paediatrician but roentgen negative are the least likely to belong to the CDH group. The following variables were tested.

1. Sex ratio
2. Birth rank
3. Fetal presentation
4. Size of the capital epiphyses

The latter variable was measured in the following way. The diameter of the epiphysis was measured on the first radiogram in which this structure was visible, in most instances 6 - 12 months after birth. The size of the epiphyses was compared between the stable and the unstable hip in unilateral cases and between the more stable and the less stable hip in bilateral cases in whom the stability was initially classified to be unequal from side to side. In bilateral cases with equal instability right hips were compared to left.

RESULTS

Sex ratio: The sex ratio in the von Rosen series was 5.2 - 1. In the next two groups the preponderance of girls was less although not significantly so (Table IV:1).

Table IV:1. Sex ratio in groups of children with varying criteria of CDH

	von Rosen	Andrén positive	Roentgen positive	Roentgen negative
Girls/Boys	5.1 - 1	4.7 - 1	3.6 - 1	1.2 - 1

The roentgen negative cases had a sex relationship which differed significantly ($P < 0.001$) from that of the other groups. These cases hardly deviated from that of the population at risk.

Birth rank: In birth rank, however, there was no significant difference between the four diagnostic groups (Table IV:2). All groups deviated from that of the population at risk ($0.05 > P > 0.02$ or better). Also in the roentgen negative cases there was a primogeniture effect.

Table IV:2. Birth ranks in children with varying criteria of CDH (per cent of group)

	von Rosen	Andrén positive	Roentgen positive	Roentgen negative
1 [1]	60	58	60	57
2	26	27	28	29
3	11	11	7	8
4 +	3	4	5	6

[1] Average for Malmö 45 per cent

Fetal presentation: The frequency of breech presentation did not differ between the von Rosen series and the two roentgen positive groups but was significantly ($0.05 > P > 0.02$) less in the roentgen negative group. However, also the roentgen negative group had a significantly ($P < 0.001$) higher frequency of breech presentation than has the population at risk (Table IV:3).

Table IV:3. Frequency of breech presentation in children with varying criteria of CDH (per cent)

	von Rosen	Andrén positive	Roentgen positive	Roentgen negative
Girls	31	23	36	13
Boys	33	29	35	27

Average for Malmö 5.7 per cent

Capital epiphyses: In unilateral cases the femoral capital epiphyses were significantly smaller in the dislocated hips and this could be demonstrated in all four diagnostic groups (Table IV:4). In bilateral cases the less stable hips had a smaller nucleus than the more stable ones in all the roentgen positive groups (Table IV:5), In the roentgen negative, however, there was no such difference.

Table IV:4. Side difference (mm) between capital epiphyses (dislocated minus stable) in children with varying criteria of unilateral CDH.

	von Rosen Right	von Rosen Left	Andrèn positive	Roentgen positive	Roentgen negative
Number	15	29	78	52	54
Average	-0.87	-1.00	-1.00	-0.62	-0.44
S.D.	1.19	1.46	1.45	1.80	1.08
	$0.02 > P > 0.01$	$P < 0.001$	$P < 0.001$	$0.02 > P > 0.01$	$0.01 > P > 0.001$

Table IV:5. Side difference (mm) between capital epiphyses (less stable minus more stable) in children with varying criteria of bilateral CDH.

	von Rosen	Andrèn positive	Roentgen positive	Roentgen negative
Number	47	98	28	8
Average	-0.64	-0.82	-0.55	-0.25
S.D.	1.52	1.33	1.12	1.04
	$0.01 > P > 0.001$	$P < 0.001$	$0.02 > P > 0.01$	$P > 0.5$

Table IV:6. Side difference (mm) between capital epiphyses (right minus left) in children with varying criteria of bilateral CDH without noticeable side difference with regard to stability.

	von Rosen	Andrèn positive	Roentgen positive	Roentgen negative
Number	20	64	27	11
Average	-0.05	0.00	-0.36	-0.45
S.D.	1.32	0.93	1.03	0.82
	$P > 0.8$	$P > 0.9$	$0.1 > P > 0.05$	$0.1 > P > 0.05$

Finally, there was no difference between right and left hips in children with bilateral CDH without side difference with regard to stability (Table IV:6).

DISCUSSION

It seems possible to draw two conclusions from these data. One is that with regard to most criteria one must assume that the cases which are treated and which would not otherwise have been detected or developed manifest dislocation also belong to the CDH group. Assuming that roentgen is the most reliable diagnostic method, especially in the hands of an experienced radiologist, there is a tendency towards a dilution of the cases with healthy children when the diagnostic criteria become less reliable. However, only the roentgen negative group deviate to any larger extent from the average. Another conclusion might be that the roentgen diagnosis adds a quality to the selection of cases for treatment which could not otherwise be obtained. This seems to hold good also for cases which may not have been radiographed exactly according to Andrén.

It has previously been demonstrated in this study that the risk involved in treatment with the von Rosen splint is negligible. Only one child developed avascular necrosis of the hip and this hip later on went on to normal. Therefore, the objections raised against the treatment and the risk involved in treating too many children could be neglected.

Is the treatment of children who otherwise would not have developed manifest CDH beneficial? Several investigators (Putti 1933, Wiberg 1939, Lloyd-Roberts 1955 and Jacobsen 1957) have implied that in osteoarthritis, particularly of the lateral type with signs of subluxation, there is a relationship to untreated CDH. Therefore, possibly, diagnosis and treatment not to mention overtreatment of CDH might decrease the incidence of osteoarthritis in the population.

PART V

JOINT LAXITY

It has in the past been demonstrated that there is a relationship between a general joint laxity and CDH. Massie and Howorth (1951) were the first to report the combination of general joint laxity and CDH. This has been confirmed by Wilkinson (1963), Carter and Wilkinson (1964[b]) and Howorth (1965). Wynne-Davies (1970) demonstrated the occurrence of joint laxity also en CDH relatives. The aim of the present study was to decide whether this could be confirmed in the Malmö CDH cases.

MATERIAL

110 of the children comprised in the follow-up study (Part II), 92 girls and 18 boys, were included in the material. For comparison, the same number of age and sex matched control children, taken from the city schools in Malmö, were investigated.

METHODS

Joint laxity was estimated by five variables (Figure V:1). It was established whether the children were able or not to:

1. Hyperextend the elbow joint more than 10^{o}.
2. Hyperextend the thumb and flex the wrist until the thumb touches the forearm
3. Dorsiflex the fingers and the wrist to make the fingers parallel to the forearm
4. Hyperextend the ankle joint more than 45^{o}.
5. Hyperextend the knee joint more than 10^{o}.

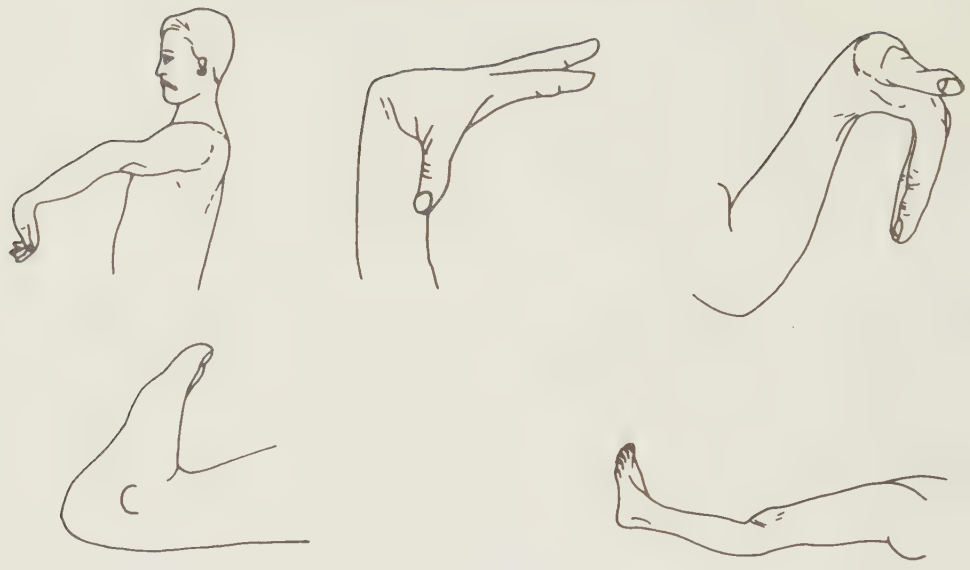

Figure V:1. Demonstration of joint laxity.

RESULTS

In all but one of the variables there was a highly significantly increased incidence of signs of joint laxity in the CDH children (Table V:1).

Table V:1. Joint laxity in patients with CDH and controls.

	CDH children		Controls		
Joint	positive	negative	positive	negative	
Thumb	65	45	52	58	$P > 0.5$
Wrist	49	61	22	88	$P < 0.001$
Elbow	40	70	6	104	$P < 0.001$
Knee	51	59	15	95	$P < 0.001$
Ankle	34	76	0	110	$P < 0.001$

DISCUSSION

The previously suggested relationship between general joint laxity and CDH is strongly supported by the findings in the children of the follow-up study.

PART VI

ALTERATION OF CONNECTIVE TISSUE IN CHILDREN WITH CONGENITAL DISLOCATION OF THE HIP [1)]

The laxity of connective tissue structures in children with CDH is well-known. Andrén (1960) demonstrated pelvic instability and several investigators (Carter and Wilkinson 1964[b] and Wynne-Davies 1970) have reported on general joint laxity in these children. A higher than expected frequency of inguinal hernia in girls with CDH is reported in Part III of the present study.

Hormonal influence of estrogens as a factor in the etiology of CDH was proposed by Andrén (1960) and Andrén and Borglin 1960 and 1961 a+b). Estrogen induced changes in the connective tissue of joint capsule and ligaments might be responsible for the increased laxity. Collagen is the major constituent of connective tissue and is responsible for the tensile strength of ligaments and joint capsule (Grant and Prockop 1972). We decided to investigate the amount of collagen and the solubility of collagen in weak organic acid in order to assess the degree of cross-linking (Jackson and Bentley 1960). As biopsies from ligament and joint capsule were not available the umbilical cord was used.

MATERIAL AND METHODS

Umbilical cords were collected from about 5,000 consecutive newborns in the maternity ward of the Malmö General Hospital. The cords were stored at $-30^{\circ}C$ until examined. Within 24 hours after delivery the children were examined by a paediatrician for clinical signs of CDH. In 39 children CDH was diagnosed and confirmed by radiological examination. Of these the umbilical cords from the 10 with the most marked instability, 7 girls and 3 boys, were selected and examined.

From the umbilical cord 5 - 6 slices of 3 - 4 mm were taken for examination. They were rinsed for blood in distilled water at a temperature of 0^{o}C. The slices were frozen in liquid nitrogen and crushed in a steel mortar. The crushed tissue was placed in a glass container with 4 times its wet-weight of 0.5 molar acetic acid. Extraction was continued for 24 hours at 4^{o}C and with constant shaking. Separation was carried out in a centrifuge at 20,000 G for 1 hour and at a temperature of 4^{o}C. The supernatant was decanted. An aliquot of each supernatant was dialyzed against distilled water for 24 hours in order to separate the smaller polypeptides that are split off when the tissue is crushed in liquid nitrogen. Using a modification of the method described by Stegeman (Pikkarainen 1968) the amount of hydroxyproline was determined in the two fractions of the supernatant and in the sediment. The amount of collagen was calculated as hydroxyproline x 7.1.

In order to examine a more homogeneous tissue we also examined the veins from the umbilical cords after careful dissection. The examination was carried out in exactly the same manner as described above for cross sections.

RESULTS

Large variations were found in both groups. The variation in collagen content was less in the veins than in the cord cross sections (Table VI:1). The collagen content was less in CDH children as compared with controls, significantly so only in the vein samples.

Extractable collagen was decreased in CDH children. The values of the non-dialyzed fraction differed significantly only in the cord cross sections. The dialyzed collagen differed between the two sets of children in the vein samples as well as in the cord sections. In the latter there was a significant difference in scatter ($P < 0.001$).

Table VI:1. Amount and solubility of collagen in umbilical cords of CDH children and controls.

Cross section of umbilical cord

	Total collagen mg/g wet weight	Extractable collagen, mg/g wet weight. non-dialyzed	dialyzed	
CDH	30.84 ± 7.48	0.198 ± 0.057	0.158 ± 0.081	n = 10
Control	38.88 ± 11.34	0.275 ± 0.041	0.197 ± 0.022	n = 10
	0.1 > P > 0.05	0.01 > P > 0.001	P > 0.1	

Vein of umbilical cord

	Total collagen mg/g wet weight	Extractable collagen, mg/g wet weight. non-dialyzed	dialyzed	
CDH	25.30 ± 3.50	0.241 ± 0.075	0.124 ± 0.056	n = 10
Control	28.75 ± 3.46	0.252 ± 0.060	0.183 ± 0.048	n = 10
	0.05 > P > 0.02	P > 0.1	0.05 > P > 0.02	

DISCUSSION

The tensile properties of connective tissue depend on collagen. The collagen content of connective tissue is decreased after administration of estrogens (Henneman 1968 and Fischer 1972). Instability of the hip in animals can be provoked by estrogen (Zaffaroni 1958, Månsson and Norberg 1961 and Gustafsson 1968 and 1975). During pregnancy the fetus is exposed to high concentrations of estrogens.

The results of the present investigation have demonstrated that the collagen content of connective tissue of children with CDH is decreased. This may explain the increased laxity of their joints and the instability of their hips. The difference in solubility indicates a difference in quality. Fetal collagen $(\alpha 1\ (III))_3$

is less soluble than adult (Miller 1973). The findings of the present study could be explained either by a different proportion of fetal and adult collagen, a decreased synthesis of collagen or difference in cross-linking in children with CDH. Our findings support the theory that CDH is a consequence of changes in the connective tissue, which is supposed to maintain the stability of the joint, rather than changes in the bony structures.

PART VII

ILLUSTRATIONS

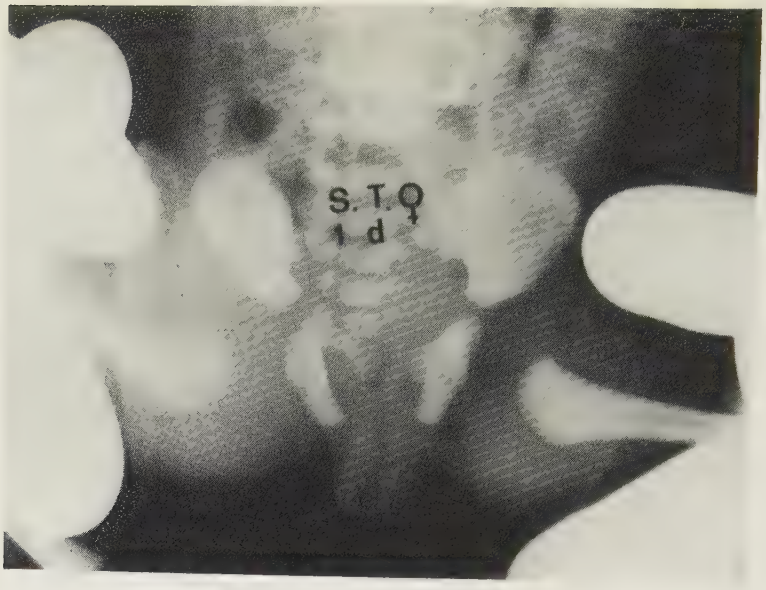

Figure VII:1. Femora pushed together.

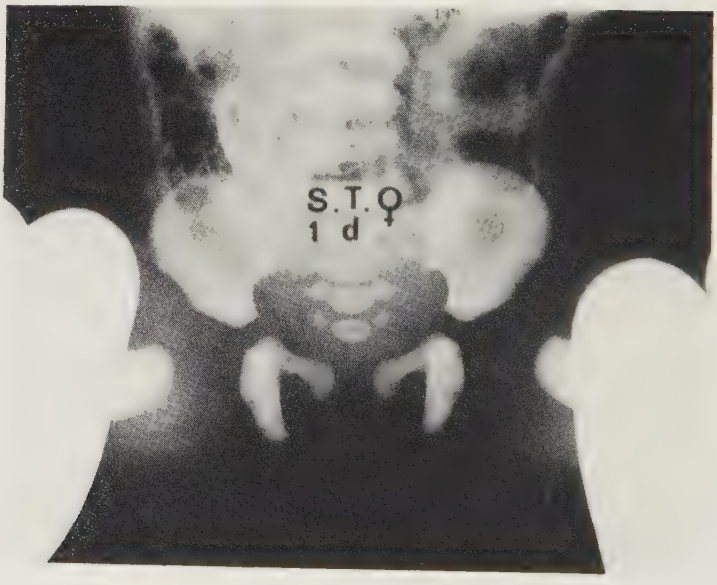

Figure VII:2. Same patient. Femora pulled apart. The symphysis has widened 2.5 millimeters. Both hips are dislocated.

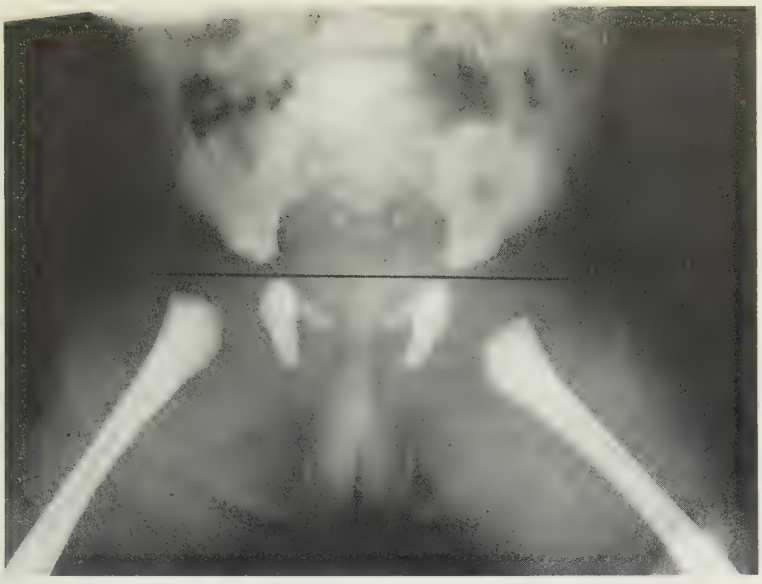

Figure VII:3. In slight abduction and outward rotation the hips are forced outwards and upwards. The right hip is dislocated.

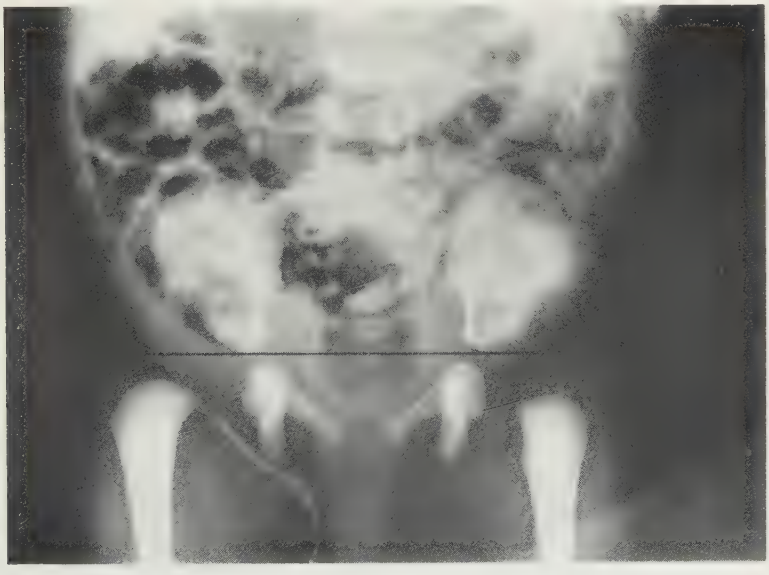

Figure VII:4. With straight legs and inward rotation the femora are forced upwards. The right hip is displaced.

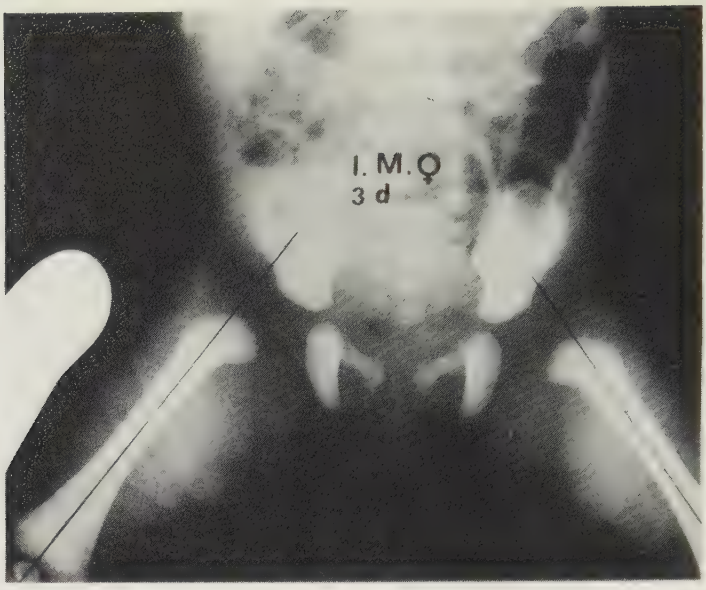

Figure VII:5. In 45 degrees of abduction and inward rotation the unstable hip is forced out of the socket. If the hip is dislocated the longitudinal axis of the femur will be projected to a point above the bony edge of the acetabulum. In this figure both hips are dislocated.

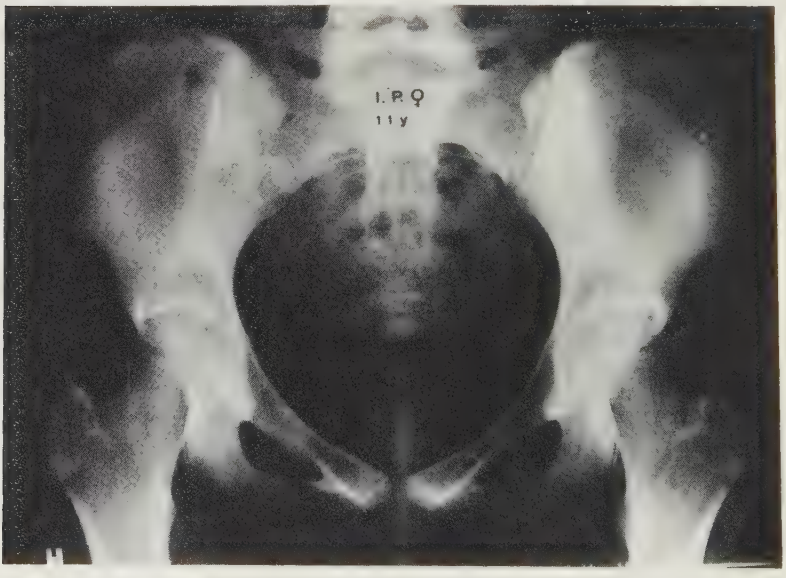

Figure VII:6. Both hips initially dislocated. Splinting was discontinued after 2 weeks and the left hip re-dislocated. After prolonged immobilization hip development was normal. This film was taken at the follow-up examination.

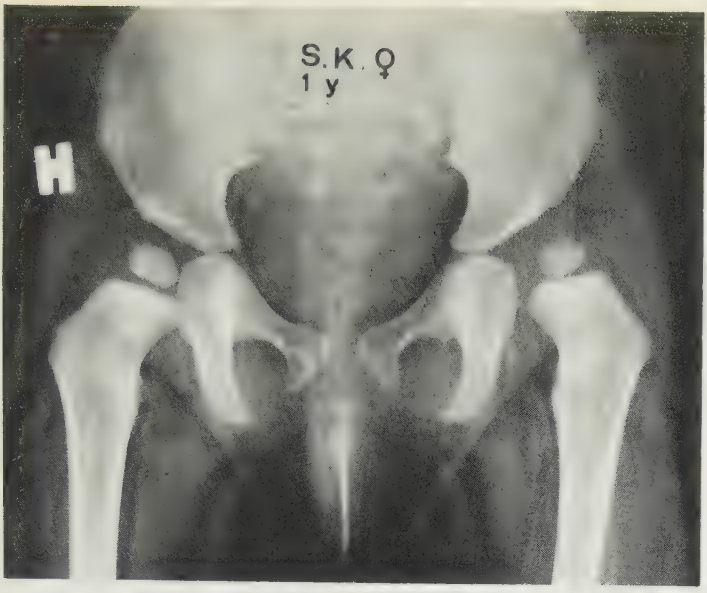

Figure VII:7. Bilateral dislocation with predominant instability on left side. Treated in the von Rosen splint for 8 weeks. At 12 months (figure) subluxation was present on the left side.

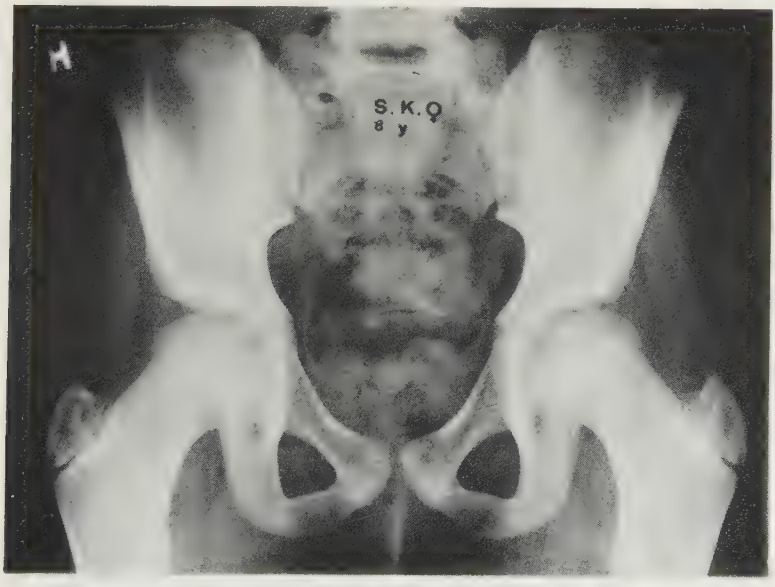

Figure VII:8. Same patient. After treatment for 3 months in a larger model of the von Rosen splint hip development was normal. Film taken at the follow-up examination.

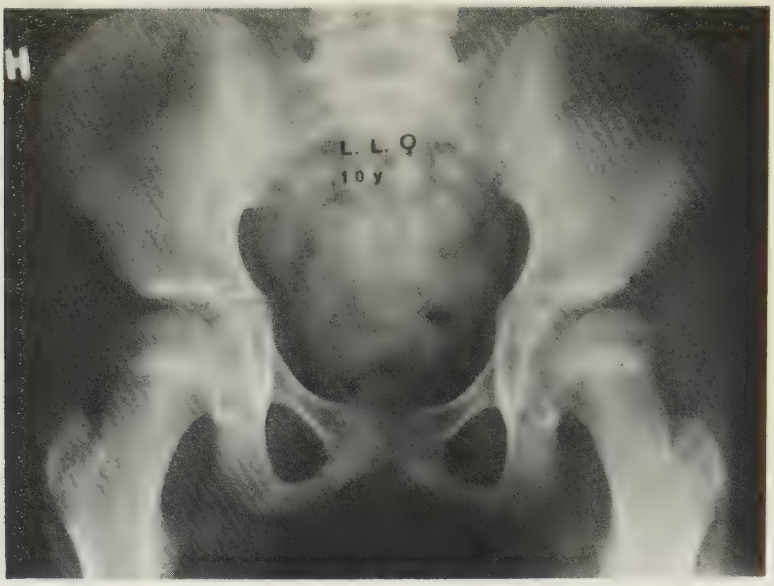

Figure VII:9. Treated without complication for a dislocated left hip. At age 10 the acetabulum on the left side is somewhat steep but the CE angle is within normal limits. No displacement of the femoral head.

Figure VII:10. Bilateral dislocation. Treated in the von Rosen splint for 10 weeks. At 2 years of age signs of avascular necrosis of the right hip (figure).

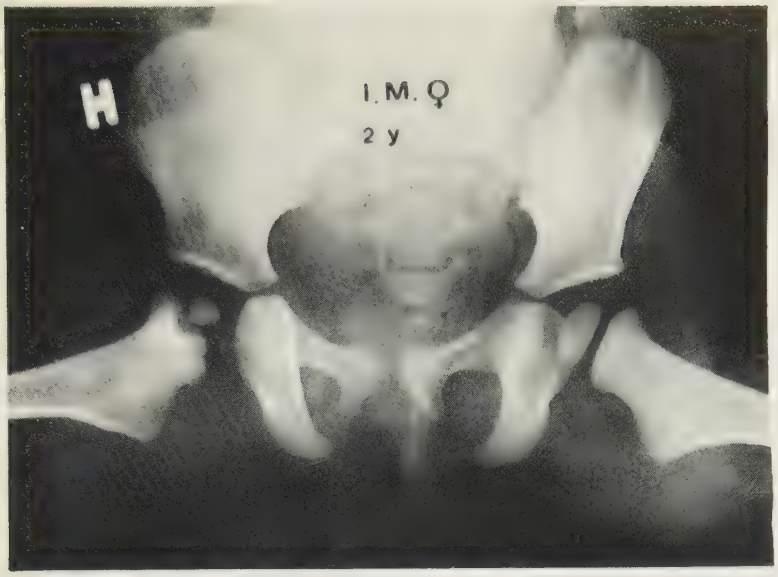

Figure VII:11. Same patient. Hips in the Lauenstein position.

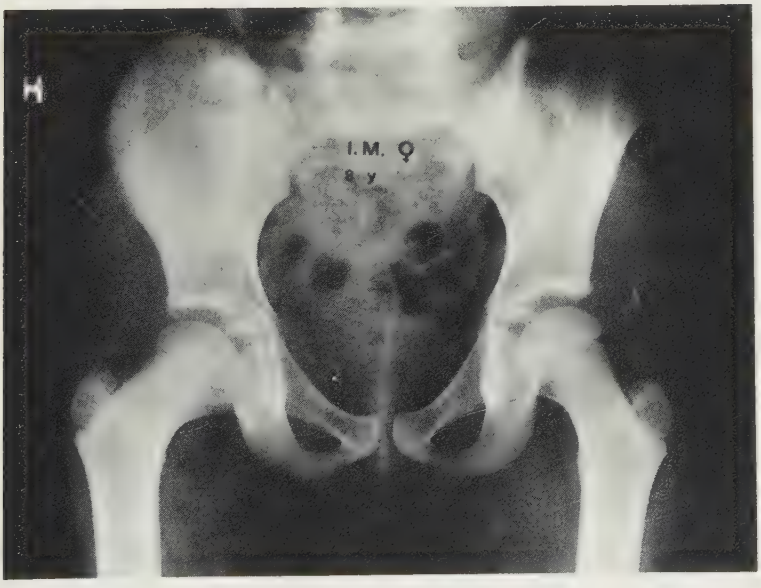

Figure VII:12. Same patient. Film taken at the follow-up examination at 8 years of age. Slight enlargement of the femoral head and neck.

GENERAL DISCUSSION

Etiology

Acetabular dysplasia as the main pathogenic mechanism, a defective "anlage", was introduced by Lorenz (1920), even if Hilgenreiner (1935) was the first to use the word dysplasia. For many years this was the predominant hypothesis. Even recently, Wynne-Davies stated that a certain number of the CDH children had dysplasia as the basic cause of CDH even if one set of the children had joint laxity. The concept of dysplasia as a primary factor in CDH provides a good excuse for failure to diagnose the condition in the newborn. Massie and Howorth (1951), Barlow (1962) and Somerville (1974) objectto this view and state that the hypothesis of dysplasia as a primary factor is incorrect, without importance for the origin of CDH and therefore no excuse for an unsuccessful diagnostic routine. In animal experiments dislocation of the hip by closed manipulation or by open surgery causes acetabular dysplasia. Dysplasia is thus secondary to dislocation. Injection of estrogens to animals produces pelvic instability.(The literature in this field was surveyed by Andrén 1962). In children with CDH an obvious pelvic instability can be demonstrated at birth and the dislocation of the hip joint is one component of this instability (Andrén 1962).

In the present study we are able to confirm the relationship between CDH and joint laxity. Joint laxity can be demonstrated in these children even when they are 10 years or older. These findings in conjunction with a suspicion that these children also have a somewhat higher incidence of hernia may point to some change in the quality and composition of connective tissue. In the present study this suspicion is confirmed in that the concentration and solubility of collagen

is different in the umbilical cord in the CDH as compared with normal children. The basic etiological back-ground to this change in the quality of the collagen is not known. It is known that estrogen acts on collagen in this way and Andrén and Borglin (1961 a+b) demonstrated defective metabolism of estrogen in CDH children. Attempts to confirm this finding have, however, failed. (Aarskog et al. 1966 and Thieme et al. 1968).

Mechanical factors interacting with joint laxity have been discussed in the past. One such factor, the breech presentation, has been confirmed in the present study. Other factors, such as ethnical differences in the handling of the newborn child, will not be commented on further.

Diagnosis

The overall situation today concerning the possibility of early diagnosis of CDH is rather confused. The experience presented in the litterature ranges from almost complete success with only an occasional case missed, to almost complete failure with the majority of cases missed. The key to this problem might well be the point in time selected for the diagnostic procedure. It appears as if those investigators who attain poor results in their diagnostic efforts either aplly their diagnostic procedure later than the first days after birth or fail to report when and in how many cases the diagnosis is delayed after that particular time period. In some instances this explanation is not valid and one might consider the possibility that experience or rather inexperience plays its part. In the present study it is shown how inexperienced physicians were given the responsibility of the diagnosis of CDH for a number of years. This, however, did not increase the incidence of missed cases but seems to have introduced an increased overdiagnosis of the condition. Possibly the attitude to the problem is of importance. If a missed case is regarded as a disastrous failure which will be investigated, this might stimulate diagnostic efforts. Another problem concerns the ethnical differences. It has been suggested that there is a second type of CDH which becomes obvious not in the newborn but later on. This means that the diagnosis is not possible to obtain in the newborn regardless of how

carefully the procedure is carried out. The few missed cases in the present study do not support this hypothesis. The reports and suggestions about this condition are mainly derived from the United Kingdom. However, it is from the United Kingdom that some of the very best and most successful applications of early diagnosis have been reported.

It should be emphasized that 100 per cent early diagnosis may not be possible to attain. A few cases are being missed even in centres with an otherwise successful organization and in most instances one can, in retrospect, find no valid explanation as to why these cases were neglected.

Does the roentgen examination of the newborn add to the diagnostic procedure? In the present study it appears as if cases with a clear radiological diagnosis are more closely related to the subset of genuine CDH. However, the technique necessary for the diagnosis includes the dislocation and reduction of the hip. A radiologist unable to do this is also unable to verify the diagnosis radiologically. In the present study the radiograms have the value of documentation. It is, therefore, not possible to claim that a greater part of these children were normal.

Treatment

The difference in opinions about the early treatment of CDH is also difficult to explain. The high incidence of complete or partial failure is surprising when compared with the good results reported by some investigators. It is true that hitherto the time of observation in most cases is fairly limited. Unlike the data in the present study, most investigators base their judgement of failure and success on radiograms taken from much younger, still rapidly growing children. Nevertheless, it seems unlikely to us that a hip which is discovered to be dislocated after 3 months of treatment was really reduced in the first place. Possibly the firm regimen instituted by von Rosen is an advantage. Nobody but experienced hospital personnel is allowed to remove the child or to put the child back in the splint. This might be an insurance against redislocation during the

first weeks of treatment. The duration of treatment, usually 3 months in this study, cannot be evaluated with respect to success or failure and could possibly be shortened in many instances. Finally, the risk of avascular necrosis may be related to the point in time in which the treatment in commenced, before or after the appearance of the abduction contracture. Also,those investigators who believe that rigid fixation of the hip of a young child is harmful might well be justified. In the von Rosen splint, a certain amount of free movement is allowed whereas in modifications of the splint with straps, the range of motion permitted to the child might be considerably limited. The position of the child in the splint may also be of importance, as we have noticed from papers describing attempts with the von Rosen treatment that the splint is improperly applied. It must therefore be suspected that some of the failures in the treatment with redislocation and "subluxation" are due to poor positioning of the child in the splint (cf. figure).

After reduction the child is immediately placed in the von Rosen splint with the hips in 90 degrees of flexion and 60-70 degrees of abduction.

Note the range of motion permitted to the child.

SUMMARY AND CONCLUSIONS

The effects of early diagnostic measures of congenital dislocation of the hip during the years 1956-1972 were investigated. The results of treatment in the von Rosen splint were examined at a follow-up study of 111 children of an average age of 10.3 years (8-16 years). Some vital statistics are presented. The large number of cases added after the early diagnosis had been introduced were investigated with regard to certain properties. The relationship between general joint laxity and congenital dislocation of the hip was investigated. The umbilical cord of children with congenital dislocation of the hip and control children was investigated with regard to the amount and solubility of collagen.

The following conclusions could be made:

1. All or almost all cases of congenital dislocation of the hip can be diagnosed in the newborn child.
2. Early treatment in the von Rosen splint leads to the development of completely normal hips.
3. Breech presentation, birth rank 1 and female preponderance are common traits in children with congenital dislocation of the hip.
4. Girls with congenital dislocation of the hip are taller and heavier than control children at the time of follow-up.
5. "Overdiagnosis" introduces a group of children with certain characteristics relating them to the CDH group.
6. The relationship between general joint laxity and congenital dislocation of the hip is confirmed.
7. The amount and solubility of collagen is decreased in the umbilical cord of children with congenital dislocation of the hip compared with controls.

REFERENCES

Aarskog, D., Stöa, K.F. and Thorsen, T. (1966) Urinary Oestrogen excretion in newborn infants with congenital dysplasia of the hip joint. Acta paediat. scand. 55:394.

American Academy of Orthopaedic Surgeons (1965) Joint Motion. Method of measuring and recording. Published by E. & S. Livingstone, Ltd., Edinburgh and London 1969.

Andersen, E. (1968) Skeletal maturation of Danish school children in relation to height, sexual development, and social conditions. Acta paediat. scand. Suppl. 185.

Andrén, L. (1960) Instability of the pubic symphysis and congenital dislocation of the hip in newborns. Acta radiol. 54:123.

Andrén, L. (1961[a]) Aetiology and diagnosis of congenital dislocation of the hip in newborns. Der Radiologe 1:89.

Andrén, L. (1961[b]) Frequency and sex distribution of congenital dislocation of the hip among breech presentations. Acta orthop. scand. 31:152.

Andrén, L. (1962) Pelvic instability in newborns with special reference to congenital dislocation of the hip and hormonal factors. Acta radiol. Suppl. 212.

Andrén, L. and Borglin, N.E. (1960) A disorder of oestrogen metabolism as a causal factor of congenital dislocation of the hip. Acta orthop. scand. 30:169.

Andrén, L. and Borglin, N.E. (1961[a]) Disturbed urinary excretion pattern of oestrogens in newborns with congenital dislocation of the hip. I. The excretion of oestrogens during the first few days of life. Acta endocr. (Kbh.) 37:423.

Andrén, L. and Borglin, N.E. (1961[b]) Disturbed urinary excretion pattern of oestrogens in newborns with congenital dislocation of the hip. II. The excretion of exogenous oestradiol-17 β . Acta endocr. (Kbh.) 37 : 427.

Andrén, L. and von Rosen, S. (1958) The diagnosis of dislocation of the hip in newborns and the primary results of immediate treatment. Acta radiol. 49: 89.

Artz, T.D., Levine, D.B., Lim, W.N., Salvati, E.A. and Wilson, Jr., P.D. (1975) Neonatal diagnosis, treatment and related factors of congenital dislocation of the hip. In preparation.

Barlow, T.G. (1962) Early diagnosis and treatment of congenital dislocation of the hip. J. Bone Jt Surg. 44-B:292.

Barlow, T.G. (1966) Early diagnosis and treatment of congenital dislocation of the hip in the newborn. Proc. roy. Soc. Med. 59:1103.

Bjerkreim, I. (1974) Congenital dislocation of the hip joint in Norway. Acta orthop. scand. Suppl. 157.

Bjerkreim, I. and van der Hagen, C.B. (1974) Congenital dislocation of the hip joint in Norway. V. Evaluation of genetic and environmental factors. Clin. genet. 5:433.

Carter, C.O. and Wilkinson, J. (1964[a]) Genetic and environmental factors in the etiology of congenital dislocation of the hip. Clin. Orthop. 33:119.

Carter, C.O. and Wilkinson, J. (1964[b]) Persistent joint laxity and congenital dislocation of the hip. J. Bone Jt Surg. 46-B:40.

Coleman, S.S. (1956) Diagnosis of congenital dysplasia of the hip in the newborn infant. J. Amer. med. Ass. 162:548.

Emnéus, H. (1966) Some new aspects on the treatment of congenital dislocation of the hip (CDH) according to Palmén-von Rosen. Acta orthop. scand. 37:311.

Emnéus, H. and Undeland, K. (1970) Två olika mönster för behandling av congenital höftledsluxation. Opusc.Med. 10:357.

Felländer, M., Gladnikoff, H. and Jacobsson, E. (1970) Instability of the hip in the newborn. Acta orthop. scand., Suppl. 130:36.

Fettweis, E. (1973) Das Hüftluxationsleiden bei in Beckenendlage geborenen Kindern. (Zur Pathogenese und Bedeutung für das Luxationsproblem). Z. Orthop.111:168.

Finlay, H.V.L., Maudsley, R.H. and Busfield, P.I. (1967) Dislocatable hip and dislocated hip in the newborn infant. Brit.med.J. 4:377.

Fischer, G.M. (1972) In vivo effects of estradiol on collagen and elastin dynamics in rat aorta. Endocrinology 91:1227.

Gustafsson, P.O. (1968) Hip dysplasia in the Greyhound: A study of estradiol induced skeletal changes. J.Amer.vet.radiol.Soc. IX:47.

Gustafsson, P.O. (1975) Estradiol induced changes in Beagles: The long term effect of prenatal and postnatal administration. Acta radiol., Suppl. 344.

Grant, M.E. and Prockop, D.J. (1972) The biosynthesis of collagen (first of three parts). New Engl.J.Med. 286:194.

Henneman, D.H. (1968) Effect of estrogen on in vivo and in vitro collagen biosynthesis and maturation in old and young female guinea pigs. Endocrinology 83:678.

Hilgenreiner, H. (1935) 10 Jahre Abduktionsschiene und Frühbehandlung der angeborenen Hüftverrenkung. Z.Orthop. 63:344.

Hindley, C.B., Filliozat, A.M., Klackenberg, G., Nicolet-Meister, D. and Sand, E.A. (1966) Differences in age of walking in five European longitudinal samples. Human Biol. 38:364.

Hippocrates 460-370 B.C. "De articuli". "The Genuine Works of Hippocrates". London 1849 (The Sydenham Society) Francis Adams II:617.

Howorth, M.B. (1965) The etiology of congenital and infantile displacement of the hip. Acta orthop.scand. 35:212.

Idelberger, K.(1951) Die Erbpathologie der sogenannten angeborenen Hüftverrenkung. Urban & Schwarzenberg, München und Berlin.

Jackson, D.S. and Bentley, J.P. (1960) On the significance of the extractable collagens. J.biophys.biochem.Cytol. 7:37.

Jakobsen, A. (1957) Vitallium mould arthroplasty for osteoarthritis of the hip joint. Oslo University Press, Oslo. The MacMillan Company, New York.

Karlberg, P., Engström, I., Lichtenstein, H., Svennberg, I. and Taranger, J. (1975) In preparation.

Knox, G. (1959) The incidence of inguinal hernia in Newcastle children. Arch. Dis.Childh. 34:482.

Lancet (1974) Editorial: Iatrogenic hip disease. Lancet, August 3, 7875:266.

Le Damany, P. (1912) La luxation congénitale de la hanche. Felix Alcan, Paris.

Lloyd-Roberts, G.C. (1955) Osteoarthritis of the hip. A study of the clinical pathology. J.Bone Jt Surg. 37-B:8.

Lorenz, A. (1920) Die sogenannte angeborene Hüftverrenkung. Ihre Pathologie und Therapie. Ferdinand Enke, Stuttgart.

MacKenzie, I.G. (1972) Congenital dislocation of the hip: The development of a regional service. J.Bone Jt Surg. 54-B:18.

MacKenzie, I.G., Seddon, H.J. and Trevor, D. (1960) Congenital dislocation of the hip. J.Bone Jt Surg. 42-B:689.

Massie, W.K. and Howorth, M.B. (1951) Congenital dislocation of the hip. II. Results of open reduction as seen in early adult period. J.Bone Jt Surg. 33-A:171.

Mears, D.C. (1974) Treatment of congenital dislocation of the hip. Lancet, September 28, 7883:780.

Miller, E.J. (1973) A review of biochemical studies on the genetically distinct collagens of the skeletal system. Clin. Orthop. 92:260.

Mitchell, G.P. (1972) Problems in the early diagnosis and management of congenital dislocation of the hip. J. Bone Jt Surg. 54-B:4.

Moore, F.H. (1974) Screening for congenital dislocation of the hip. J. Irish med. Ass. 67:104.

Månsson, J. and Norberg, I. (1961) Höftledsdysplasi hos hund. Medl. bl. Sver. Vet. För. 1 :330.

Ortolani, M. (1937) Un segno poco noto e sua importanza per la diagnosi precoce di prelussazione congenita dell´anca. Pediatria 45:129.

Ortolani, M. (1951) Frühdiagnose und Frühbehandlung der angeborenen Hüftgelenksverrenkung. Kinderärztliche Praxis 19:404.

Palmén, K. (1953) Congenital höftledsluxation: Diagnos och behandling hos nyfödda. Svenska Läkartidningen 50:1433.

Palmén, K. (1957) Dysplasia coxae congenita. Svenska Läkartidningen 54:1706.

Palmén, K. (1961) Preluxation of the hip joint. Acta paediat. Suppl.. 129.

Palmén, K. (1970) Preluxation of the hip in the newborn: The diagnostic work in Sweden during the years 1953-1966. Acta orthop. scand. Suppl. 130:8.

Palmén, K. and von Rosen, S. (1975) Late diagnosis congenital dislocation of the hip. To be published in Acta orthop. scand.

Pikkarainen, J. (1968) The molecular structures of vertebrate skin collagen. A comparative study. Acta physiol. scand. 309:22.

Ponseti, I.V. (1966) Non-surgical treatment of congenital dislocation of the hip. J. Bone Jt Surg. 48-A:1392.

Putti, V. (1927) Per la cura precoce della lussazione congenita dell´anca. Arch. ital. Chir. 18:653.

Putti, V. (1933) Early treatment of congenital dislocation of the hip. J. Bone Jt Surg. 15:16.

Record, R.G. and Edwards, J.H. (1958) Environmental influences related to the aethiology of congenital dislocation of the hip. Brit. J. prev. soc. Med. 12:8

von Rosen, S. (1956) Early diagnosis and treatment of congenital dislocation of the hip joint. Acta orthop. scand. 26:136.

von Rosen, S. (1959) Early diagnosis and early treatment of congenital hip luxation. Acta orthop. scand. 29:164.

von Rosen, S. (1961) Our further experience in the treatment of congenital hip joint dislocation in newborns. Acta orthop. scand. 31:242.

von Rosen, S. (1968) Further experience with congenital dislocation of the hip in the newborn. J. Bone Jt Surg. 50-B:538.

von Rosen, S. (1970) Instability of the hip in the newborn. Acta orthop. scand. Suppl. 130:13.

Roser, W. (1879) Über angeborene Hüftverrenkung. Langenbeck´s Archiv für klinische Chirurgie 24:309.

Severin, E. (1941) Congenital dislocation of the hip joint. Late results of closed reduction and arthrographic studies of recent cases. Acta chir. scand. Suppl. 63.

Severin, E. (1956) Frekvens av luxatio coxae congenita och pes equino-varus i Sverige. Nordisk Medicin 55:221.

Snedecor, G.W. and Cochran, W.G. (1967) Statistical Methods. The Iowa State University Press.

Somerville, E.W. (1974) The nature of the congenitally dislocated hip. Proc. roy.Soc.Med. 67:1169.

Thieme, W.T., Wynne-Davies, R., Blair, H.A.F., Bell, E.T. and Loraine, J.A. (1968) Clinical examination and urinary oestrogen assays in newborn children with congenital dislocation of the hip. J.Bone Jt Surg. 50-B:546.

Walker, G. (1971) Problems in early recognition of congenital hip dislocation. Brit.med.J. 3:147.

Weissman, S.L. and Salama, R. (1966) Treatment of congenital dislocation of the hip in the newborn infant. J.Bone Jt Surg. 48-A:1319.

Weissman, S.L. and Salama, R. (1969) Treatment of congenital dislocation of the hip in the newborn infant. J.Bone Jt Surg. 51-A:601.

Wiberg, G. (1939) Studies on dysplastic acetabula and congenital subluxation of the hip joint. Acta chir.scand., Suppl. 58.

Wiberg, G. (1953) Shelf operation in congenital dysplasia of the acetabulum and in subluxation and dislocation of the hip. J.Bone Jt Surg. 35-A:65.

Wilkinson, J. (1963) Prime factors in the etiology of congenital dislocation of the hip. J.Bone Jt Surg. 45-B:268.

Wilkinson, J. (1972) A post-natal survey for congenital displacement of the hip. J.Bone Jt Surg. 54-B:40.

Wilkinson, J. and Carter, C. (1960) Congenital dislocation of the hip.The results of conservative treatment. J.Bone Jt Surg. 42-B:669.

Williamson, J. (1972) Difficulties of early diagnosis and treatment of congenital dislocation of the hip in Northern Ireland. J. Bone Jt Surg. 54-B:13.

Wynne-Davies, R. (1970) Acetabular dysplasia and familial joint laxity: Two etiological factors in congenital dislocation of the hip. J.Bone Jt Surg. 52-B:704.

Zaffaroni, A. (1958) Hormones and meat production from livestock. Recent Progr.Hormone Res. 14:213.